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ORIGINAL ARTICLE

A normal screening ultrasound does not provide complete reassurance in infants at risk of hip dysplasia; further follow-up is required

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Abstract

Background Screening for developmental hip dysplasia (DDH) continues to evolve with the use of ultrasound (US) in either selective or universal screening methods. The possibility of delayed evidence of DDH, and thus the need for radiographic follow-up at a later stage of development have been suggested by some authors.

Aims The aim of this review was to evaluate the number of patients in our hospital network with a normal screening US at 6 weeks with evidence of DDH at the time of radiographic review at 6 months. Secondary aim; to determine the outcomes for these patients.

Methods A retrospective review was done to infants undergoing DDH ultrasound screening between January and December 2015. Initial US and radiographs at 6 months were reviewed. Patients with normal screening US who had subsequent radiographs were included for analysis.

Results In total, there were 829 patients included for analysis. Sixty-three patients (8%) had evidence of DDH at 6 months, representing 34% of all DDH diagnoses for the study period. Five of the 63 patients were lost to follow-up. The remaining 58 babies were treated in Boston bracing. Four patients with evidence of persistent DDH were referred for tertiary review. The osteotomy rate in the radiograph diagnosed group was 2%, versus 6% and 3% in the unstable and US diagnosed groups, respectively.

Conclusion Eight percent of patients with a normal screening US had evidence of DDH at time of radiograph at 6 months, reflecting 34% of all our DDH cases for the year. Based on these findings, patients in our hospital network undergo radiographic evaluation at 6 months even if the initial screening US is normal.

Keywords Developmental dysplasia of the hip · Radiograph · Screening · Ultrasound

Introduction

Developmental dysplasia of the hip (DDH) is one of the most common developmental abnormalities and includes a spectrum of anatomic abnormalities from dysplastic shallow acetabular development to complete hip dislocation. A paper from our unit reported an incidence of DDH of 6.73 per 1000 live births making it a significant condition for screening and early treatment [1]. Other authors report incidence of 8 per 1000 live births [2]. Patients with early diagnosis of DDH and intervention in early infancy demonstrate more normal growth and are less likely to require operative intervention than those diagnosed later [3].

There is as yet no consensus on imaging or clinical practice guidelines for the early detection and non-operative management of DDH. Screening guidelines range from clinical screening only to selective or universal ultrasound screening [4, 5]. The subsequent follow-up of infants with normal ultrasound also varies, with some centres using radiographic follow-up [1, 6, 7], with other centres not carrying out radiographic follow-up of at-risk infants [8–10].

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We hypothesised that a proportion of patients presenting for US screening for DDH, with normal screening ultrasound, would have evidence of dysplasia on radiograph at 6 months, and a proportion of these late diagnosed patients would have persistent dysplasia requiring surgery.

Aim

The aim of this review was to evaluate the number of babies in a single hospital network with clinically stable hips and a normal ultrasound (US) at 6 weeks with evidence of DDH on radiograph at 6 months. A secondary aim was to determine the number of these radiograph-diagnosed infants requiring surgery for persistent DDH after bracing.

Methods

We performed a retrospective review of patients presenting for DDH ultrasound screening at the study hospital between January 2015 and December 2015. Medical charts and radiographic databases were searched for data including sex, risk factors and other indications for referral. We defined major risk factors as breech presentation or positive first-degree family history.

Ultrasound and radiograph reports were reviewed, for findings consistent with DDH. The initial ultrasound findings and initial radiograph findings were recorded. Where reports were non-specific, measurements were taken from radiographs, using the PACS software (McKesson Enterprise Medical Imaging, Change Healthcare) including the acetabular index [11] (Fig. 1.) An AI of more than 30° was considered abnormal; patients with AI of 27° or more were referred for orthopaedic review. In the following review, patients, with AI 27° or more, were considered abnormal and treated if there were clinical findings suggestive of DDH such as asymmetric hip abduction. The total number of DDH diagnoses for the year was also determined by clinic logbooks for harnessing and clinic referrals for dislocation.

Patients with radiographic diagnosed DDH were treated in a Boston brace until normal radiographs and clinical examination and were followed until walking age. Patients with persistent DDH after bracing were referred to a tertiary Paediatric Orthopaedic centre for further management.

Logistic regression analyses were performed to determine the risk of DDH for gender, presentation at birth and family history of DDH. Odds ratios (OR), 95% confidence intervals (CI) and p values were determined, with a 5% level of significance assumed to be statistically significant. The statistical analysis was performed using SAS Version 9.4.

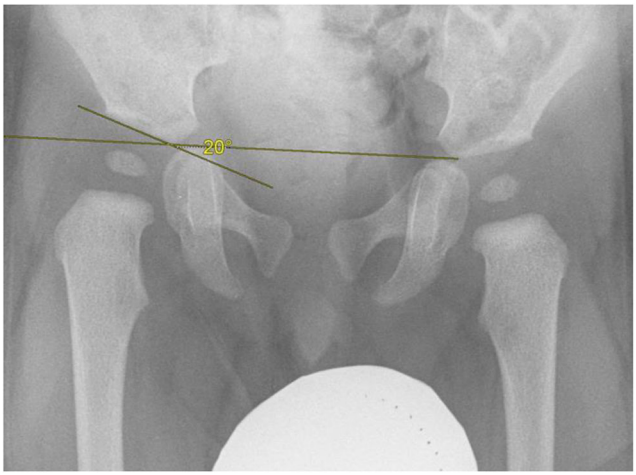

Fig. 1 The acetabular index (AI) as subtended by Hilgenreiner's line and acetabular roof

Inclusion criteria

Records of all patients attending the DDH ultrasound clinic during the study period were checked. Our service operates a selective US screening programme for DDH. All patients with a normal screening ultrasound were included in the study. Patients with an immature (Graf IIa) hip on initial ultrasound were included if subsequent US at 3 months showed normal hips without harnessing treatment.

Exclusion criteria

Patients were excluded if there was clinical evidence of instability by a documented Ortolani or Barlow manoeuvre or if harnessing was commenced for any period prior to initial radiograph. Patients without follow-up radiograph at 6 months were also excluded. Patients with Graf IIa hips who did not attend for repeat US at 3 months were excluded.

Results

There were 997 patients attending for US screening during the study period. Sixteen of the US examinations were done for babies who had already a harness applied for clinical instability and were excluded. A further 45 had no radiographic follow-up at 6 months and were excluded. Two patients with Graf IIa hips at 6 weeks did not attend for US at 3 months and were also excluded. There was evidence of DDH at the time of initial US in 33 patients and these were treated and excluded. There were 779 babies with normal findings on initial US. One hundred and twenty four infants had immature (Graf IIa) hips on initial US, and repeat US at 3 months showed mature (Graf I) hips in 50 of these patients. In total, there were 829 patients with a normal initial or 3-month US included for follow-up (see Fig. 2).

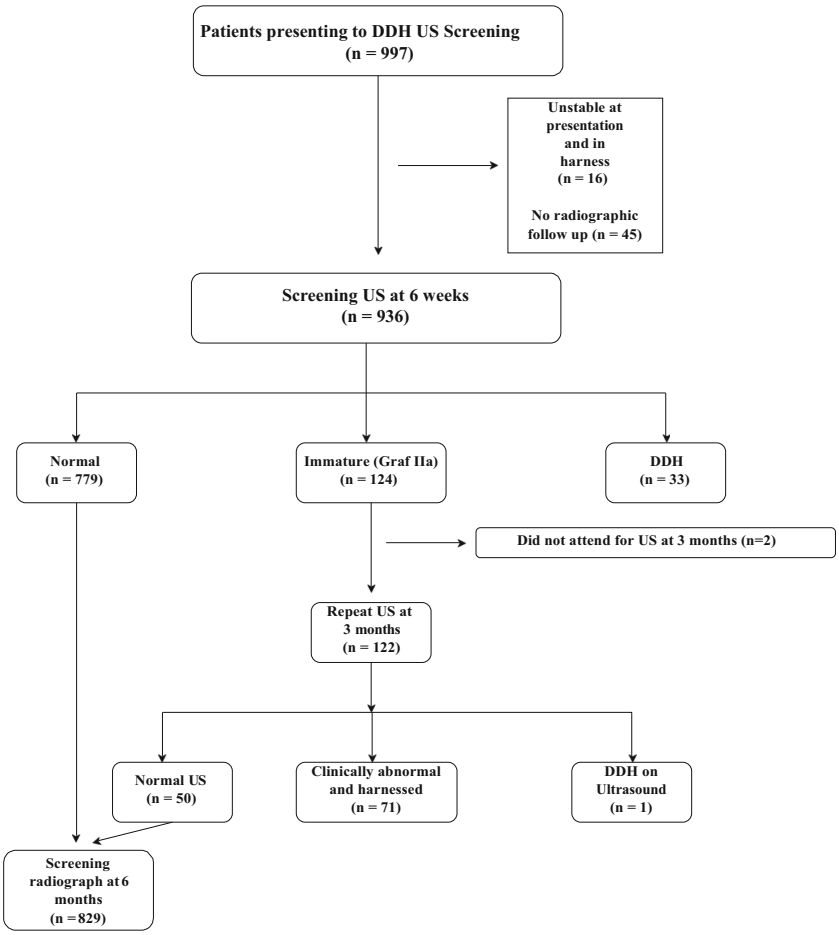

Fig. 2 Selection pathway for included population ($N = 997$)

Primary outcome

Of the included patients, 429 (52%) were female and 400 (48%) were male. The number of patients with major risk factors was 571 (68%): breech presentation in 245 (29%), 320 (38%) babies with a positive first-degree family history and 6 (1%) with both. 'Clicky hips' was another common indication for referral (17%). All referral indications are shown in Table 1.

There were 63 patients with abnormal radiographs at 6 months, giving an overall radiographic pick up rate of 8% (Fig. 3).

The logistic regression analyses found that female gender was a strong risk factor for DDH in those with normal ultrasound examinations (OR = 3.9, 95% CI = 2.1–7.4, $p < 0.001$). Family history of DDH showed an increased risk for DDH but this was not statistically significant (see Table 2). There were

Table 1 Demographics and risk factors of screening population ($N = 829$)

Characteristic		*n* (%)
Gender	Female	429 (52%)
	Male	400 (48%)
Indication	Breech presentation	245 (29%)
	First degree family history of DDH	320 (38%)
	Breech and first degree family history of DDH	6 (1%)
Other referral	'Clicky hip'	141 (17%)
	Asymmetric skin creases	11 (1%)
	Abnormal examination	41 (5%)
	Other non-first degree family history	21 (3%)
	Unknown	4 (1%)
	Other	41 (5%)

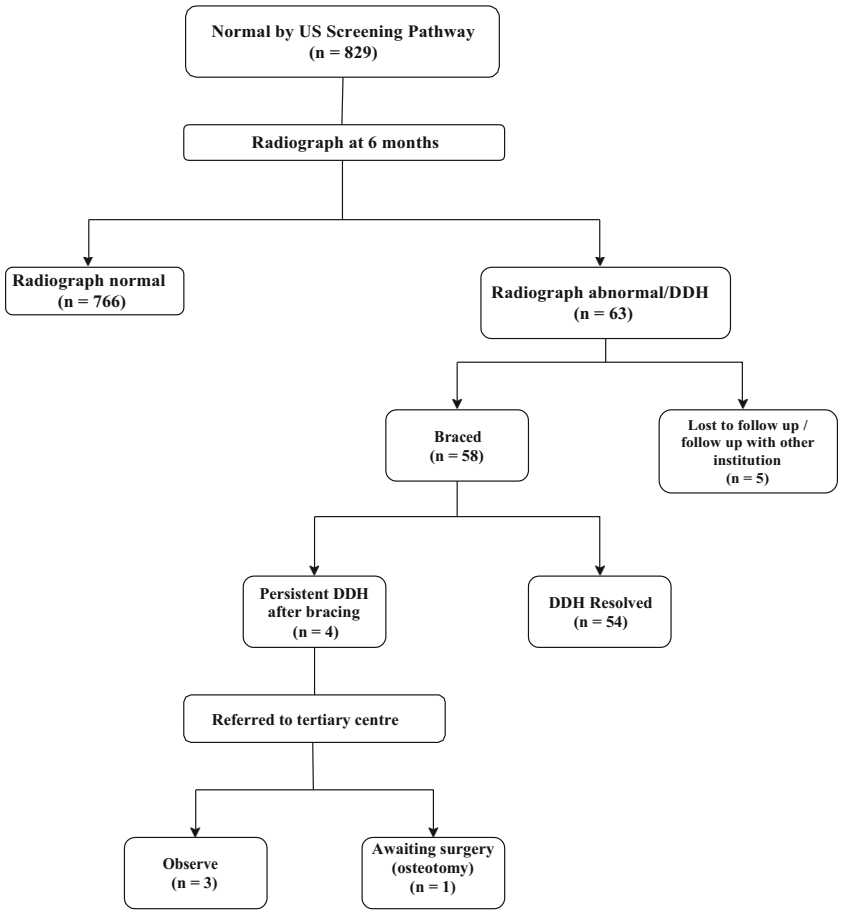

Fig. 3 Outcomes of study population with normal ultrasound ($N = 829$)

184 DDH diagnoses for the year, 63 (34%) were diagnosed on radiograph at 6 months. Sixteen infants had unstable hips at birth, and a further 168 infants were diagnosed on screening.

Secondary outcomes

Five of these 63 patients were lost to follow-up. All remaining infants with DDH diagnosed at 6 months were treated in Boston brace with regular follow-up until examination and radiographic parameters normalised and until walking age. Four infants had persistent dysplasia by walking age and were referred for tertiary paediatric orthopaedic review. Surgery was performed in one patient (2%) in the radiograph-diagnosed group (Tables 3 and 4.

Table 2 Risk of late diagnosed DDH in those with a normal ultrasound examination

Risk factor	OR (95% CI)	*P* value
Female gender	3.9 (2.1–7.4)	< 0.001
Family history of DDH	1.3 (0.7–2.1)	0.390
Breech presentation at birth	0.8 (0.4–1.4)	0.389
Family history + breech	2.5 (0.3–21.4)	0.414

Discussion

DDH screening continues to evolve. US screening is widely accepted internationally; however, US techniques and population selection for screening vary. Follow-up after initial normal US is also not agreed upon. Some authors have suggested abolishing the radiographic follow-up in infants with normal US screening whilst other authors have shown evidence to support follow-up with X-ray [6, 7].

Our aim was to determine the rate of late diagnosis of DDH on 6-month radiograph in infants with previously normal screening ultrasound.

We found an 8% rate of late detection of DDH, which contributed 34% of the overall DDH diagnoses for the year.

The corrective osteotomy rate was 3% after US diagnosis, 6% in the clinically diagnosed group and 2% in the radiograph diagnosis group. This suggests that the patients with normal US, who were diagnosed on subsequent radiograph, were at the milder end of the pathological spectrum. This would be similar to the findings shown in other studies, where infants with normal US and subsequent radiographic evidence of DDH, normalised without intervention [8, 9, 12].

The natural history of DDH is not fully understood, making it a difficult condition for which to develop a universally

Table 3 Demographics of babies with radiographic-diagnosed DDH ($N = 63$)

Characteristic		*n* (%)
Gender	Female	50 (79%)
	Male	13 (21%)
Indication	Breech presentation	15 (24%)
	First degree family history of DDH	27 (43%)
	Breech and first-degree family history of DDH	1 (2%)
	Clicky hip(s)	9 (14%)
	Abnormal examination	6 (9%)
	Other	5 (8%)
Side of DDH	Left	19 (30%)
	Right	23 (37%)
	Bilateral	21 (33%)

accepted screening programme. DDH screening practices still vary internationally and authors differ on recommendations. Imrie et al. found that, in breech infants with normal initial US, there was a 29% radiographic diagnosis rate at 6 months, and they advocated radiographic follow-up of hips with normal US [6].

Sarkissian looked at a subset of infants with abnormal US at 6 weeks which normalised by the 3-month US and found these had 25% rate of dysplasia on X-ray [7]. This would be similar to our cohort of infants with initial Graf IIA hips, included in the study population.

In contrast to both Imrie and Sarkissian's high late pick up rate, other authors report rates from 1 to 5%, even in patients with positive family history [8, 9, 12]. All of these cases resolved without treatment by 30 months. One of the authors even followed infants with high normal values until their hips were well within normal range [12]. This could reflect the less severe disease spectrum in the sample group. These studies also had small sample sizes: 89–181 patients [8, 9, 12]. Price et al. reported 11,000 patients screened with US and follow-up radiograph at 5 months. In their study, patients with milder dysplasia were followed with imaging only, and orthopaedic review was sought only where persistent changes were seen on imaging. Of the infants referred, 4% required osteotomy [13]. This pathway differed from that at our institution, where orthopaedic referral is made once there is evidence of dysplasia on imaging. This is accompanied by both a higher diagnosis rate and lower surgical rate.

In considering results, important factors to note are US reporting methods, and the inherent learning curve in applying US techniques. Different methods of US techniques have been used in the reporting of DDH [14]. The Graf US method is widely adopted across Europe and has a known inherent learning curve, as is common in US techniques. Price et al. studied over 11,000 patients with risk factors for DDH [13]. They initially utilised the Graf method of US screening. Their rationale for follow-up radiographs was to cover for the inherent learning curve in the US method. Our study utilised the Graf method of reporting, and as with any US technique, reporting reproducibility must be considered, though the Graf method has shown adequate interobserver agreement for use in screening [15].

In recommending radiographic follow-up of at-risk infants, there is concern regarding radiation exposure. Our estimated radiation dose was 0.02 mSv at exposure factors of 62 kV and 2.5 mAs. Other authors estimated radiation exposure and justified doses based on radiographic diagnostic rates [6].

Comparison in the reported literature is difficult, because of the variation in US methods used, the combination of methods in some institutions and the varied risk groups for which outcomes have been published. Thus, in formulating a screening programme, consideration must be given to these differences in the reported literature.

Strengths in our study include the large sample size, inclusion of all risk factor groups for screening with subgroup analysis and single orthopaedic consultant follow-up for standardised management protocol and clinical diagnosis.

Table 4 Characteristics of X-ray diagnosed patients with persistent DDH

Gender	Side	Risk factor/indication	First ultrasound	Outcome
Female	Bilateral	Family history	Normal	Observe
Female	Bilateral	Breech	Normal	For osteotomy
Female	Left	Clicky hip	Normal	Observe
Female	Left	Other	Normal	Observe

Limitations to our study include variability due to multiple reporters of radiographs from different institutions within the network, as well as the inherent learning curve associated with the Graf ultrasonic method by multiple sonographers, though within the same specialist unit. Additionally, there was no control group for treatment of the radiograph-diagnosed group, since all such patients underwent bracing.

Conclusion

Our overall late radiographic diagnosis rate in at-risk infants was 8%, reflecting 34% of all our cases of DDH for the year. Based on these findings, patients in our hospital network undergo radiographic evaluation at 6 months even if the initial screening US is normal.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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